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FAMOCA

FAMILY ONLINE COUNSELING FOR FAMILIES WITH PARENTAL CANCER

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Abstract

Parental cancer is a huge challenge to affected patients and their families. Often, affected parents of minor children are totally overwhelmed by the new diagnosis and need support, particularly concerning the communication with their spouse and children.

Therefore, the aim of this research project was the evaluation of the feasibility and the effects of a web-based counseling intervention during the first months of cancer treatment, aimed at the improvement of coping at the level of children, couples, and the family.

Publication 1 describes the set-up of the web-based program in a family affected by parental cancer immediately after diagnosis. The web-based program consisted of four modules of four weeks each, individualized for each family member. The publication demonstrates that web-based counseling is a feasible option for all family members with parental cancer.

Publication 2 focuses on the feasibility of the web-based program and its effects on participating children. The children's first stage adjustment to parental cancer did not show detrimental patterns. The major "lesson learned" in this setting was the challenge to contact and motivate families in need, not only starting, but also keeping up on the program.

Publication 3 examined the feasibility and the effects on psychological adjustment focusing on challenges of the couple, using psychometric testing in patients and their partners before and after initial therapy, both on the program and on the control group. No comparisons to controls were possible. Signs of anxiety were present in patients and partners in the first phase of cancer treatment. A slight improvement in anxiety and psychological wellbeing was shown at follow-up.

Taken together, feasibility was shown, but the effects on participants in this web-based counseling intervention could not be compared to controls due to low numbers in both groups.

Introduction

Over the past 20 years, there has been growing awareness and evidence that cancer is a disease affecting a whole system, such as a family, and not just a single patient's diagnosis. A recent cancer diagnosis marks a major transition in a family's life, challenging the family's stability at every stage, inner-family relationships, and quality of life of the individual family members. Rearrangement of roles and responsibilities, renegotiation of plans about the future of the individuals and the family as a whole, adjustment to loss of functioning, fear of recurrence, burden of care in a possible palliative situation, and preparation for death are enormous challenges to the family system. Emotional responses of individual family members affected by parental cancer are inter-dependent and mutually influencing (Pitceathly & Maguire, 2003; Schmitt et al., 2008). Newly diagnosed parental cancer at the same time affects the individual (patient, partner, and children), the couple, and the parent-child relationship – thus initiating heterogeneous coping patterns. Lewis (Lewis, 2010) summarizes several "*stuck points*" experienced by families affected by parental cancer: First, the lack of parental knowledge, the lack of skills, and the high levels of distress impeding the support of dependent children. Second, the uncertainty and the shortfalls in spouses on how to respond supportively to the patient's distress. Third, the patients' and spouses cancer associated psychosocial morbidity interfering with family functioning and especially the children's functioning. Forth, the tension put on the marriage by the newly diagnosed cancer negatively affects the household's functioning. Fifth, the use of coping behavior that may not be responsive to changing demands from the cancer-affected individual – therefore being ineffective to reduce the cancer-related burden. Hence, it can easily be argued that interventions to improve adjustment to parental cancer should include spouse-related, parent-child related and family-related interventions.

As of now, intervention studies aimed at the improvement of child or family adjustment to parental cancer were conducted as face-to-face interventions. Interestingly, the internet as a commonly used source of information by patients and caregivers was neglected for a certain time as a supporting resource, particularly as a means for conducting interventions and gathering new evidence. However, the internet as a source of delivering psychosocial interventions in psychooncology has recently become evident (Leykin et al., 2011). Among other sources, the internet may be used

and may be favorable in family-based interventions, as information on cancer and coping strategies can be provided in a standardized and age-specific way. Furthermore, all inputs and activities can be used individually; irrespective of time and place, and independent of other family members, as well as adapted to the family's schedule.

Aims and theoretical background

The randomized and controlled trial primarily aimed at the evaluation of the efficacy of a web-based counseling intervention during the first 5 months of cancer treatment for the improvement of coping at the level of minor children, couples and the family.

Secondary aims were the identification of clinical predictors predicting benefits in individuals and families, and the identification of clinical predictors prognosticating failure or continuing adjustment problems. The primary hypothesis was that this web-based intervention would be superior to usual care. If moderate effect-sizes (similar to the much more demanding face-to-face interventions) were shown, the evidence for the establishment of low-threshold contact and entry to web-based psychooncological support would be strengthened. If predictors of failure or success were to be identified, the disposition to web-based or personal psychooncology support and therapy could be put on a rational basis. Therefore, the support of more appropriate treatment decisions (web-based counseling vs. more intensive family therapy/other approaches) was the ultimate goal of the project.

Impact of cancer on relationship / marriage

A recent meta-analysis based on studies using clinical interviews concludes that the combined prevalence estimate of mental health conditions in patients in acute cancer care is 32% (Singer, Das-Munshi, & Brahler, 2010). Similarly, Pitceathly and Maguire (Pitceathly & Maguire, 2003) found 20-30% of partners to be probable cases of psychiatric morbidity based on studies using self-report questionnaires, mounting up to 30-50% in the case of advanced disease. Several studies have shown that females are more likely to be emotionally distressed and develop affective disorders, as compared to males (Baider, Koch, Esacson, & Kaplan De-Nour, 1998; Hagedoorn, Buunk, Kuijer, Wobbes, & Sanderman, 2000). Besides an avoidant coping style (Manne, Ostroff, Winkel, Grana, & Fox, 2005) and negative appraisal (Kershaw et al.,

2008) as intrapersonal variables, several interpersonal factors have been identified to predict couple adjustment to cancer: availability of mutual support and confidants (Baider & Denour, 1999; Ell, Nishimoto, Mantell, & Hamovitch, 1988; Walker, 1997), marital satisfaction currently and prior to cancer (Pitceathly & Maguire, 2003), and communication skills (Pitceathly & Maguire, 2003).

Taken together, cancer may disrupt relationship functioning on several levels. Patient and partner are individually challenged to cope with the physical and emotional consequences of cancer and to deal with the emotional response and coping efforts of the partner (Hagedoorn, Kreicbergs, & Appel, 2011). Adjustment difficulties result in the disruption of intimacy and sexuality, less cohesion and more communication problems, less mutual support and greater marital conflict (Baider et al., 1998; Baik & Adams, 2011; Lambert & Girgis, 2017; Lambert et al., 2013; Saita, Acquati, & Molgora, 2016; Zimmermann, 2015).

Impact of parental cancer on children and adolescents

Children with parents suffering from cancer have a higher risk of developing behavioral problems or mental disorders (Rutter, 1966; Visser et al., 2005). Overall, about 25-30% of the children of early stage cancer patients are identified with internalizing, externalizing, and emotional problems (Birenbaum, Yancey, Phillips, Chand, & Huster, 1998; Heiney et al., 1997; G.A. Huizinga et al., 2011; Siegel et al., 1992; Visser et al., 2005). This is supported by the German COSIP study which showed a 31% (norm population 16%) prevalence of child behavior problems (Child Behavior Check List CBCL) in children of parents affected by cancer (Romer & Haagen, 2007). Adolescents self-report more problems than their parents note (Heiney et al., 1997), which might be explained by the less open expression of feelings and worries, in order to protect the family (Davey, Gulish, Askew, Godette, & Childs, 2005). In general, girls seem to be more vulnerable than boys (Gazendam-Donofrio et al., 2007; Huizinga et al., 2005; Welch, Wadsworth, & Compas, 1996). Coping strategies and family functioning seem to be predictors of the child's psychological well-being. Adolescents are more likely to develop emotional and behavioral symptoms compared with younger children. However, the heterogeneity of these findings and of the study designs does not allow for strong conclusions to be drawn (Faccio, Ferrari, & Pravettoni, 2018).

While adjustment in children improves over the first year after a parental cancer diagnosis, children who show initial problems remain vulnerable over the first year (Visser et al., 2007) and unfortunately long term data are lacking, unfortunately.

Medical parameters, such as duration and stage as well as prognosis of the parental illness have little or no impact on psychological problems of children (Compas et al., 1994; Howes, Hoke, Winterbottom, & Delafield, 1994; Huizinga, Visser, Van der Graaf, Hoekstra, & Hoekstra-Weebers, 2005; Lewis, Hammond, & Woods, 1993; Visser et al., 2005). Instead, a child's adjustment to parental cancer is *moderated* by the child's developmental stage, gender (girls having more problems), and the supporting network (G A Huizinga et al., 2011). For the development of a family-based counseling program, these variables, which mediating the child's response to parental cancer, are of particular importance, as they can be approached by interventions. Among these factors, the child's appraisal of the parental illness, the availability of coping strategies, parental psychological functioning, marital satisfaction, parenting skills, family functioning and family communication have been found to foster child adjustment to parental illness (G A Huizinga et al., 2011).

Depending on previous history, stage of development, type of communication within the family, relationship satisfaction of the parents, and - most importantly - psychological functioning of the parents, the children's adjustment may vary greatly (Visser, Huizinga, Van der Graaf, Hoekstra, & Hoekstra-Weebers, 2004). Risk factors for maladaptive adjustment in children were shown to be parental factors, but also factors of the family system and external factors, such as socio-economic status, or level of education. Among the parental factors, higher levels of depression in both parents (Edwards et al., 2008; Grabiak, Bender, & Puskar, 2007; Lewis & Darby, 2003; Sigal, Perry, Robbins, Gagne, & Nassif, 2003; Visser et al., 2004; Watson et al., 2006) were shown to be of utmost importance. Among the family factors, adjustment patterns were repeatedly shown to be crucial. Chaotic or less structured adjustment (G. A. Huizinga et al., 2005), a low extent of open communication (Watson et al., 2006), enmeshment and low family affective involvement (Watson et al., 2006), low family cohesion (Huizinga, van der Graaf, Visser, Dijkstra, & Hoekstra-Weebers, 2003; Watson et al., 2006), as well as a general dysfunctionality in family relationships and in the adaptation to changes of roles (Visser et al., 2004) were the main factors

identified. Other factors associated with maladaptive adjustment were maternal cancer in girls (Visser et al., 2004), cumulative traumatic stress (Fischer & Riedesser, 1999), single parents affected by cancer (Visser et al., 2004), and – generally – a lower socio-economic status and lower levels of education (Egle, Hoffmann, & Steffens, 1997; Sattler & Font, 2018; Woolfenden et al., 2015). The findings of a recent study exploiting semi-structured interviews with 29 mothers diagnosed with cancer indicate a high amount and diversity of stressors and strains for the ill and healthy parent and for their children, respectively. At the same time, parents make use of diverse resources and coping strategies on external, family or intrapersonal level. The assessment of strains and resources may be an important indicator for the support needs of families when a mother has cancer. Enhancing and activating resources and coping strategies may help the families to manage the situation better and may prevent maladjustment in the family members (Inhestern & Bergelt, 2018). This study relied on the Family Adjustment and Adaptation Response (FAAR) Model, which that hypothesizes that the balance between strains and resources of families affected by cancer can be an important indicator on the families' adjustment to the situation.

Couple- and family-based interventions

Couple counseling

Experiencing cancer challenges a couple's communication skills, roles and responsibilities (Carlson, Bultz, Speca, & St-Pierre, 2000). Thus, a number of couple based interventions have been developed to provide emotional support to the patient and his/her partner (some validated in a German speaking population (Heinrichs & Zimmermann, 2008). Most of the intervention programs involve the partner actively (Baik & Adams, 2011). Depending on the stage of the illness, they focus on relational competences, such as communication skills, decision-making and problem solving to improve communication, reciprocal understanding, and intimacy and reduction of illness-related distress (Baik & Adams, 2011). At a later stage of disease, other factors, such as existential concerns, caregiving, and anticipated loss may play a more important role (Zaider & Kissane, 2009).

Couple's interventions not only improve marital functioning, but also reduce depressive symptoms in patients, as well as symptoms of pain. They were shown to be more

efficacious than patient-only psychosocial intervention and usual care, respectively (Arden-Close & McGrath, 2017; Cano & Tankha, 2018; Helgeson, Jakubiak, Van Vleet, & Zajdel, 2018; Jacobs et al., 2017; Kiecolt-Glaser & Wilson, 2017; Martire & Helgeson, 2017; Martire, Schulz, Helgeson, Small, & Saghafi, 2010).

Family counseling

Several counseling concepts for families newly affected by parental cancer have been developed and evaluated. Interventions tended to focus primarily on the parents, while children were rarely systematically included.

The Enhancing Connections Programme (EC) (Lewis, Casey, Brandt, Shands, & Zahlis, 2006) is a cancer parenting program that focuses on five factors known to affect the quality of a child's adjustment to parental cancer: parental mood and anxiety; parenting skills; parenting confidence; the quality of the parent-child relationship and the child's cancer-related concerns. The program has been evaluated for efficacy in a RCT with families with children between 8-12 years and showed significant improvements in parent and child outcomes (mood, behavioral-emotional functioning) after the intervention up to 12 months post-baseline (unpublished data) (Lewis, 2011). The German COSIP study (Romer & Haagen, 2007; Romer, Kühne, Bergelt, & Moller, 2011) is a preventive intervention program for children of somatically ill parents. Its objectives, subdivided in family-, parent- and children-systems, focus on the encouragement of open communication within the family, the flexible treatment of individual needs of all family members, and the enhancement of coping strategies in children and parents. As part of the multi-centric cooperative project "Kinder körperlich kranker Eltern", the COSIP manual was evaluated for feasibility and acceptance. Results were promising, but most interventions were not planned to be web-based, and research ended 12 years ago (Inhestern, Geertz, Schulz-Kindermann, & Bergelt, 2018; Romer et al., 2007; Thastum, Munch-Hansen, Wiell, & Romer, 2006).

Internet-based counseling approaches in psychooncology

The so far discussed intervention studies were all planned, set up, and conducted as face-to-face interventions. Only recently, the research field of "web-based

interventions" focusing on the treatment of psychological problems and disorders in general, and in cancer patients in specific, has become overt.

Cancer patients and caregivers commonly use the Internet as a source of information (Kinnane & Milne, 2010). Amongst others, the availability of information at any hour and anonymity are highly appreciated (Strecher, 2007). For counseling, the internet overcomes some of the known barriers in attending to psycho-oncologic support (stigma and privacy concerns, geographical distance form providers, time constraints to adhere to additional appointments during office hours) (Leykin et al., 2011). In addition, taking the side of the provider, the Internet is a time- and cost-effective method to convey interventions improving psychological adaptation to cancer; given the shortage of mental health services available to cancer patients, which is still a concern in Switzerland, internet interventions have a potential impact from a clinical oncology and a public health perspective.

E-mail therapy, self-help, prevention, counseling or psychoeducational programs are only a few examples for these new forms of therapy. A specific combination of these different intervention types are so called "minimal-contact" or "guided self-help" therapies, which deliver interactive treatment components presented via web pages, and provide ongoing support using e-mail (Berger & Andersson, 2009; Newman, Erickson, Przeworski, & Dzus, 2003). Several controlled trials in the field of anxiety and mood disorders and behavioral medicine have provided evidence for the effectiveness of web-based therapy (Barak, Hen, Boniel-Nissim, & Shapira, 2008; Barkmann, Romer, Watson, & Schulte-Markwort, 2007; Berger, Hohl, & Caspar, 2009; Spek et al., 2007).

In cancer patients, studies have shown that online support groups *without* professional moderation and self-guided online interventions *without* interactive tools to link the patient and the provider, are less effective than interventions which include components such as monitoring symptoms, guided homework or practice, individualized tracking and customized feedback (e.g. via email) (Høybye et al., 2010; Kroenke et al., 2010; Owen et al., 2005; Ritterband et al., 2011). Given an interactive format, web-based interventions have shown comparable effect sizes as face-to-face interventions. For example, a couple-based study on sexual adjustment showed Cohen's d of .35 for both, web-based and face-to-face counseling (Schover et al.,

2011), with a potential limitation for cancer populations with a high degree of psychopathology (David, Schlenker, Prudlo, & Larbig, 2011). Web-based interventions in cancer patients were limited, as of yet, to individuals, couples, or groups of affected individuals. To our knowledge, there were no studies evaluating the effects of minimal contact intervention on entire families affected by parental cancer at the start of the project. Web-based counseling was provided - but not scientifically evaluated - in children and adolescents of parents with cancer (www.kinder-krebskranker-eltern.de). Users were mostly between 12 and 23 years old. They seemed to search support mostly in order to interact with an independent trained therapist on their individual family situation. In additional chat room exchanges with other children affected by parental cancer, the experience was the focus on stressors and mutual support (Trabert & Zimmermann, 2011).

Methods

Taking into account a moderate effect size, as published in the literature, 90 families with minor children were planned to be included in to the randomized controlled trial. Specifically, families with minor children confronted with a cancer diagnosis of a parent - conveyed less than a month before inclusion - were to be randomized to either the "Web-based counseling for families with parental cancer program" or to a control condition providing a reader for parents and children based on existing information booklets ("usual care"). Figure 1 summarizes the design of the study.

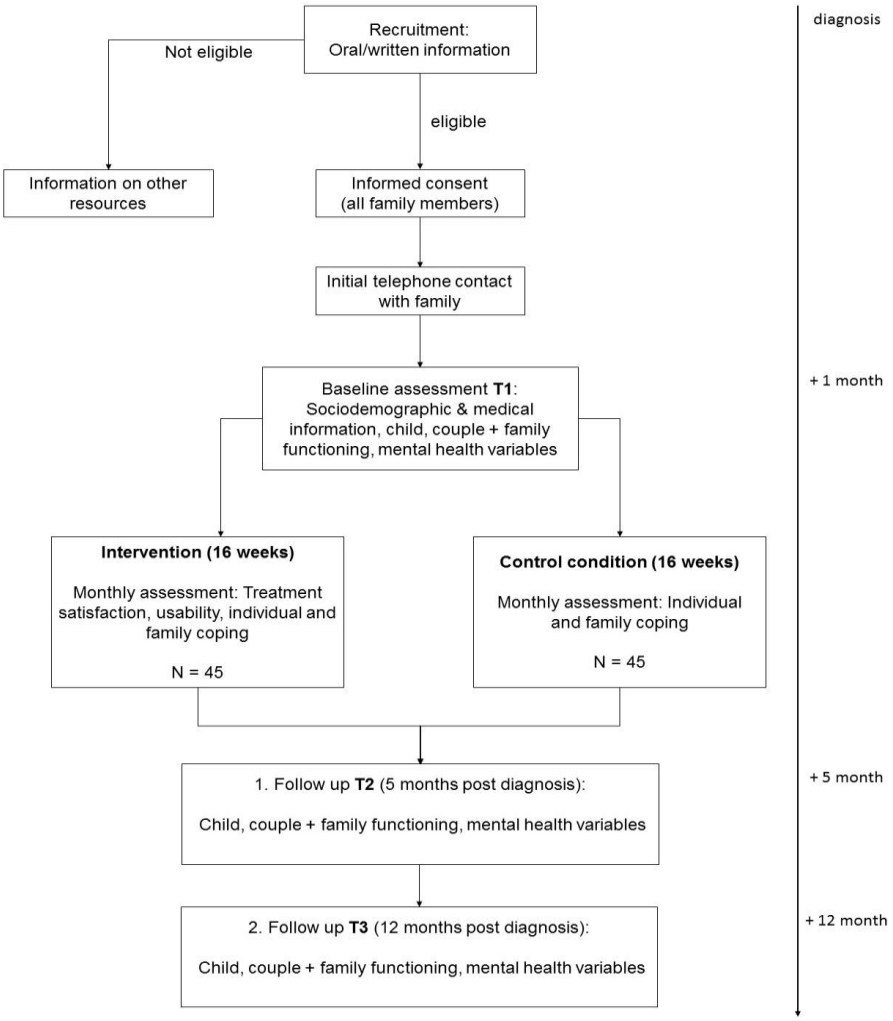

Figure 1. *Design of the Study*

The intervention consisted of four modules “recognition”, “adaption”, “coping”, and “change”. Parents had to complete two sessions per module each month, whereas children and adolescents only had one per month. The program contained separate sections for parents and children with teenagers working on their own and parents working with their younger children. Sections for parents were not accessible for adolescents and vice versa, whereas the children’s section was open to all family members. Besides the delivery of written information, the internet format allowed for a multimedia-approach including interactive coping exercises, educational videos about cancer and its treatment, or downloads of relaxation exercises. Parents and teenagers had the possibility to visit a forum (separate for parents and adolescents) to exchange

experiences or to discuss self-selected topics with others. All participants had the opportunity to directly contact the study team regarding any concerns. Monthly assessments on treatment satisfaction and short evaluation of family coping and individual wellbeing were conducted. A trained psycho-oncologist or a psychologist in training under supervision accompanied every family until completion of the program.

Parents, adolescents, and children had age-specific and adapted contents per module. Every four weeks, a new module was activated.

Primary outcome was family adjustment (cohesion, flexibility and communication). Secondary outcomes were child adjustment (behavioral problems and quality of life), couple adjustment (communication, intimacy and conflict), and parental mental health and coping. Furthermore, feasibility was to be assessed for this minimal contact intervention by analyzing inclusion, dropout, and qualitative feedback by participants.

Summary of Results

The following section contains a short overview of the main results of each article. Article 1 was published 2018 in Psychooncology (Bingisser et al., 2018). Article 2 and 3 are submitted to a peer-reviewed journal. All articles are listed in the appendix, where further information on methods and detailed prescription of the results may be found.

Publication 1

Web-based counseling for families with parental cancer (Bingisser et al., 2018)

This article evaluated a web-based counseling program designed to support all family members and described its effects on a family after the diagnosis of breast cancer in a mother of 2 children. The research question focused on the qualitative feedback by participants and the data on the use of the program.

The family logged into the program 33 times and the combined use was 20.6 hours, the mother and patient spending over 10 hours online, while the partner spent 4 hours, and the children over 3 hours online.

Regarding the qualitative feedback, overall ratings of the program’s user-friendliness, as judged by the parents and the adolescent, were good. All participants expressed a

desire for more audio and video files. The patient reported subjective improvements in anxiety and in cohesion, flexibility, communication, and satisfaction at the end of the program, while the partner indicated psychosocial distress at the end of the program.

Feasibility was shown using the time spent online and the subjective effects on coping, anxiety, and family cohesion.

Publication 2

Web-based counseling for families with parental cancer: Baseline findings and lessons learned (Denzinger et al., accepted)

The research question was the quality of life and emotional-behavioral wellbeing in children affected by parental cancer, and the family communication and satisfaction and feedback on the web-based program. This article described results in all 22 families participating in the FAMOCA study. Results have shown that children's first stage adjustment to parental cancer did not show detrimental patterns.

Of 35 direct referrals, 22 families with a total of 36 children were enrolled. Of the 22 enrolled families, seven were in the control and 15 in the intervention group. Nine families completed all assessments, three completed baseline and one-year assessments, and ten families only completed baseline assessments. No significant differences were found comparing dropouts with completers regarding demographic or diagnostic characteristics. Reasons for not completing all modules were lack of time, loss of interest, and death of parent.

Children between three and six years reported a normal quality of life, while their parents reported sub-threshold scores. Children between seven and seventeen also reported a normal quality of life, very much similar to the younger children, except for "emotional wellbeing" which was lower. Children generally showed higher than normal values in the subscales *hyperactivity/inattention* and *peer relationship problems*, whereas parents rated their teens higher than self-scores in the subscale *emotional symptoms*.

82% of all parents and 88% of all adolescents rated communication levels as high to very high. 24% of parents and 50% of adolescents rated family satisfaction levels

between high and very high while 44% of parents and 25% of adolescents rated satisfaction level between low and very low.

On average, participants spent 32 minutes on the website. In general, families appreciated the variety of provided information and the freedom to work independently with the program, as well as the monthly phone contact with the psychologist. Children and adolescents liked the active parts of the program (e.g. storybook). Most families criticized that FAMOCA provided too much text to read; some patients mentioned concentration problems due to cancer treatment and children felt challenged due to their daily schedule.

Taken together, these results showed a good adjustment of children to their parent's new cancer diagnosis. Families rated the impact of FAMOCA to be moderate regarding the adjustment process.

Publication 3

Feasibility of minimal contact interventions is limited in couples affected by a new cancer diagnosis. (Bingisser et al., submitted)

The research question was to determine whether couples affected by a cancer diagnosis would participate during the first five months of cancer therapy in the described minimal contact intervention, and to report on psychological adjustment before and after the minimal contact intervention.

Of 292 eligible patients, 33 patients contacted the study team, and 13 patients could not be included for the following reasons: unwillingness, time constraints, palliative treatment, and other reasons. Finally, 20 patients and 14 partners were included. All patients had one to three underage children. Dropout in patients was 50%. Anxiety was pronounced at diagnosis in patients and partners, but decreased at 16 weeks of follow- up. Optimism was subdued at diagnosis, but increased during follow up.

Due to the low numbers, patients and partners were pooled from intervention and control groups. There were no differences in demographic or outcome variables between the groups. Depression scores showed sub-threshold values in the majority of patients without significant change after 16 weeks. Anxiety levels were above the

cut-off scores in the majority of patients and partners, with significant decrease in patients after 16 weeks.

Scores for pessimism showed mildly elevated values for the majority of patients and partners without significant change after 16 weeks. *Scores for optimism* showed subdued values for the majority of patients and partners without significant increase in patients after 16 weeks.

Quality of marital relationship showed intermediate values for the majority of patients and partners at the time of diagnosis, without significant change after 16 weeks, the only exception being a significant difference in the rating of conflict behavior between patients and partners.

Patients showed high scores for the following coping styles: active coping, use of emotional support, instrumental support, positive reframing, planning, and acceptance.

The lessons learned emphasizes the challenge to reach families at need. FAMOCA was appreciated as an additional source of information and support in this mostly highly functioning population.

Discussion

The aim of our research project was the evaluation of the feasibility and efficacy of a web-based counseling intervention during the first 5 months of treatment for the improvement of coping at the level of minor children, couples and the family.

Several results deserve discussion: First, participating families were – in spite of a pronounced anxiety – highly functional at the start and showed no significant deterioration of coping, family adjustment, anxiety, and depression in the course of the study. Second, the quality of marital relationship showed intermediate values for the majority of patients and partners at the time of diagnosis, without significant change after 16 weeks. Third, FAMOCA, as a minimal contact intervention, was shown to be feasible. However, feasibility was hampered by low recruitment and high dropout. Therefore, a certain selection of families has to be taken into account. Families adhering to the program spent a considerable time online. It is highly unlikely that more than 20 hours of face-to-face counseling can be provided to an entire family within the first months of diagnosis. The program can be used irrespective of time and place, and participants can spend as much time as needed online and freely choose their topics of interest (Andrews, Cuijpers, Craske, McEvoy, & Titov, 2010). Some discrepancies between reported conflict behavior and intimacy between patient and partner might illustrate both an advantage and a disadvantage of online programs. Psychometric data could be gathered and analyzed at regular intervals by such programs. This is hardly the case in face-to-face counseling. So, minimal contact programs could support the “objective observation” of couples and families. If, e.g., the program detected discrepant ratings, an online-alarm could be conveyed to a psychooncologist. On the other hand, if such findings are only assessed by the program and not discussed professionally, they may be missed and further therapy may be refrained from.

Another advantage of online programs is the support of families with less obvious need for intensive psychooncological therapy. The low-threshold support was appreciated by the majority, and the specific information about cancer and family effects were among the denoted needs.

Though feasibility was shown, no comparison between intervention and usual care was possible due to low recruitment and high dropout rates.

How do our results compare with the literature? For cancer patients, studies show that online support groups *without* professional moderation and self-guided online interventions *without* interactive tools to link the patient and the provider, are less effective than interventions which include components such as monitoring symptoms, guided homework or practice, individualized tracking and customized feedback (e.g. over email) (Høybye et al., 2010; Kroenke et al., 2010; Owen et al., 2005; Ritterband et al., 2011). Given an interactive format, web-based interventions have shown comparable effect sizes to face-to-face interventions: for example, a couple-based study on sexual adjustment showed Cohen's d of .35 for both, web-based and face-to-face counseling (Schover et al., 2011), with a potential limitation for cancer populations with a high degree of psychopathology (David et al., 2011). Web-based interventions for cancer patients so far have been provided in single, couple and group format while to our knowledge there are no studies evaluating its effectiveness for families affected by parental cancer. For children and adolescents of parents with cancer web-based counseling is provided but not scientifically evaluated, e.g. as in the case of the German website www.kinder-krebskranker-eltern.de. Its users are mostly between 12 and 23 years old and search support to talk about their family situation with an independent trained therapist and in the chat room exchange with other children affected by parental cancer where they talk about stressors and support each other (G A Huizinga et al., 2011; Trabert & Zimmermann, 2011). Similar to our data, 70% of all families showed a normal adjustment (Osborn, 2007; Singer et al., 2010; Visser et al., 2004). However, no studies have analyzed family functioning prior to diagnosis. Therefore, one of the possible inclusion biases could be the overrepresentation of highly functioning families. This seems a common limitation of all studies in this field. Other lessons taken from the literature include the discussion on "low-threshold support". Generally, caregivers believe that they offer low-threshold support. However, if threshold was really low, inclusion rates should be higher. Further, most programs provide information and support on open communication and positive coping strategies. This support, however, will largely depend on the background of the respective family, such as the level of education, the biopsychosocial model of illness, and on functional pre-existing coping strategies. Each individual family member might need individual support and differentiated interventions – just as the family as a whole. For example being that mothers of smaller children were emotionally more distressed

due to their concerns about the attachment to their children compared to parents of older children who were more concerned about practical issues, such as taking care for the children after day-care or household duties as major challenges for the families (Inhestern et al., 2018). Unfortunately, no single study was conducted in the same manner as FAMOCA. Therefore, direct comparisons are not possible. However, lessons from the existing literature on qualitative and semi-structured interviews were implemented. The need for professional support of communicating between family members (Semple & McCaughan, 2013), but also of communicating with the extended family or institutions, has been shown multiple times, and minimal contact interventions should implement these needs.

Strengths and Limitations

One of the strengths of the study was the implementation of a minimal contact program based on the pre-existing literature. Therefore, the needs of families were met and could be documented. FAMOCA is still the only study focusing on all family members, including small children, using the minimal contact method. Another strength is the independence of time and place - inherent to online programs - as well as the psychometric data provided. These data might well be used in clinical practice as an additional source of information on the families' wellbeing.

Limitations are multiple, unfortunately due to the small sample size. Efficacy could not be shown, as the group sizes were too small for statistic comparisons. Therefore, the scientific evidence on the impact of the FAMOCA program could not be brought forward.

Generalizability is also limited due to a possible selection bias. Our sample mainly consisted of families with no migration background, good general resources and a high income and education level. Most of the couples were married and usually the mother suffered from breast cancer. Interestingly, previous research had similar limitations (Moyer, Sohl, Knapp-Oliver, & Schneider, 2009).

As single parenthood is a risk factor for the development mental disorders in children (Weitoft, Hjern, Haglund, & Rosen, 2003), this vulnerable population should be studied. Unfortunately, no single parents could be included – and by means of the method, only patients with curable disease were included.

Additionally, the program was only available in German. Therefore, about 30% of all eligible patients (average percentage of non-German speaking patients in our hospital) could not be included.

Due to a possible over-recruitment of highly functional families, it remains unclear what effects one could expect in families with lower psychosocial functioning.

Clinical Implications and future research

The major clinical implication is the feasibility of this online program. Obviously, there is a need to reach out to all family members. In every-day clinical life, patients are screened for distress and subsequently (if individual distress exceeds a certain limit) supported by face-to-face counseling. If no evidence for distress in other family members is detected, they are not contacted by the caregivers and may lack support thereafter. Another implication is the need regarding information and support in the fields of informing children about the newly diagnosed cancer in a parent. Interestingly, supporting material, such as printouts, books, and homepages are readily available. However, patients may lack information or even the possibility to get such information, particularly tailored to the needs of the individual family member. Therefore, online-programs such as FAMOCA offer the opportunity to a much broader audience to get information and support in the often catastrophical event of a new cancer diagnosis in a parent.

Future research has to focus on finding better access to the population at need – e.g. families who suffer psychosocially from cancer, families with low resources, and families with a higher pre-existing psychosocial burden and lower income, respectively. Further, it is of interest to find out which families could possibly get enhanced benefit from face-to-face counseling. More research is needed to determine what elements future interventions should contain to meet the needs of the children, the parents and the family as a whole. It remains unclear, how to contact families with lower psychosocial functioning, as they were mostly lost to inclusion in the existing literature.

In spite of one of the big advantages, namely the standardization of information given using such minimal contact interventions, there is no good evidence on the content of such information (Li & Loke, 2014). Therefore, not only expert opinion, but also opinion of patients and family members should be sought in order to refine such programs and

find evidence for the best content for each individual, e.g. pre-school children, school-age children, adolescents, and adults – ideally tailored according to psychosocial background and the biopsychosocial model of illness.

Taken together, more studies comparisons between minimal contact and face-to-face counseling should be attempted, in order to gain evidence on the differences in outcomes, the differential indications, and the identification of prognostic factors for the benefits of each program. It is highly likely that a combination of both methods could provide better outcomes than each method by itself. Therefore, combinations of face-to-face and online counseling for families affected by parental cancer should be studied in the future.

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Appendix

RUNNING TITLE: Web-based counseling for families

Web-based counseling for families with parental cancer: a case report

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Key words

Parental cancer, web based counseling, minimal contact, coping, family adjustment

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RUNNING TITLE: Web-based counseling for families

Introduction

Cancer not only affects patients, but has a massive impact on families (1). The stress caused by the disease can lead to additional difficulties in coping, emotional distress, and anxiety in patients and their families (2). Counseling protocols for families with parental cancer have been published, primarily for face-to-face settings (3). Alternatives include web-based programs specifically designed to promote the development of coping skills (4). However, these interventions are designed for patients, not for other family members.

We therefore evaluated a web-based counseling program designed to support all family members. This case report describes its effects on a family after the diagnosis of breast cancer in a mother of two children.

Material and Methods

Design

FAMOCA (Family online counseling for families with parental cancer) is a web-based program aiming to improve psychological adjustment in families of newly diagnosed cancer patients. Based on the principles of cognitive-behavioral therapy, it focuses on psycho-educative elements, and aims to enhance open communication, affective involvement, family and relationship cohesion, problem-solving skills, and adaptive coping at the level of the individual, the couple, and the family. We evaluated the program in a prospective, randomized, controlled intervention study that was approved by the Ethics Board of Basel (EKB).

Recruitment

Families were recruited by means of leaflets distributed in waiting rooms and articles published in magazines, websites, and online forums. Inclusion criteria were a recent diagnosis of first parental cancer with an expected progression-free survival of at least 12 months and least one child between 3 and 18 years in the family.

Assessments

Assessments comprised psychometric tests of family functioning, parental mental health, and parental coping at baseline (t1), at the end of the 16-week program (t2),

and after one year (t3). Families randomized to the intervention group had access to FAMOCA.

Intervention

FAMOCA is based on the "minimal contact" concept: family members participated autonomously in the online program and received individual monthly feedback from a psychologist (5). The website for the intervention group (www.famoca.ch) provides individual support for each family member, with age-specific sites for children, adolescents, and parents. The program consists of four successive modules, each lasting four weeks (see supplemental appendix table 1 to 3).

Outcome Measures

Data were analyzed using an open source analytics platform (www.piwik.org). We used the Hospital Anxiety and Depression Scale (HADS) (6) to assess depression and anxiety, the Partnership Questionnaire (PFB; subscales: *conflict behavior, intimacy, and mutuality)* (7) to assess quality of the partnership, and the Family Adaption and Cohesion scale (FACES; subscales: *family cohesion, family flexibility, family communication,* and *family satisfaction*) (8) to assess the overall cohesion of the family (see supplemental appendix table 4).

Monthly feedback on coping and well-being from each family member separately, as well as for the family as a whole was assessed using Likert scales (0–5 for coping, and 0–10 for well-being). Open questions, such as "how well did you feel last month" completed the assessment.

Case Report

The 47-year-old female patient H.D., living with her husband F.D. (46 years), and two sons (N.D. 11 years, and T.D. 8 years), was diagnosed with breast cancer. She underwent 16 weeks of neoadjuvant chemotherapy, subsequent surgical removal of the tumor, and standard postoperative radiotherapy.

H.D. came across FAMOCA during her online research and registered three weeks after her diagnosis. She was worried that her children were afraid she was going to die, and felt that the module "understanding what's going on" helped her and her husband to plan how to talk to their sons and what information to provide.

After thirteen weeks, weakened by chemotherapy and an influenza infection, H.D. developed depressive symptoms. She judged the psycho-educative elements and coping exercises offered by the program to be a good support during this time. Furthermore, she felt that her partner responded with increased emotional closeness. In the monthly mails to her psychologist, and at the end of the program (week 16), H.D. stated that the family was coping well, and she felt that the program had helped them to cope.

Partner Report

F.D. worked full time and supported the family during his wife's chemotherapy by taking on more childcare and household responsibilities. He felt rather insecure in talking to his children about his wife's cancer. The information provided on communication skills therefore helped him considerably. He also benefited from the training provided in coping skills and stated that the family members' well-being improved during the first three months.

Children's Reports

N.D. (11 years) was assigned to the adolescent's section at his mother's request. However, he spent 37% of his time in the children's section. He described the information on cancer as helpful. He did not work through all modules. He found that his family coped well with the situation and attributed this to the program. He indicated that his life had not changed due to his mother's illness with respect to family relations, friendship, school, hobbies, his own feelings, or his everyday life stress.

T.D. (8 years) logged into the children's section with his mother and was mostly supervised by his mother. She claimed that T.D. benefited most from the children's stories, the handicraft instructions, and the diary. She felt that the program helped T.D. to cope with the situation.

Outcomes

Online Sessions

The family logged into the program 33 times and used it for 20.6 hours. The patient (H.D.) spent 10.1 hours (16 sessions) online, viewed 327 pages, and downloaded 53. She spent 1.6 hours in the children's sections. The husband (F.D.) spent 4.1 hours (7

sessions) online, viewed 142 pages, and downloaded 16. He spent 1.3 hours in the children's sections. The older son (N.D.) spent 3.2 hours (7 sessions) online, viewed 108 pages, and downloaded 26. The younger son (T.D.), spent 3.2 hours (three sessions) online, viewed 79 pages, and downloaded 21.

Monthly feedback

For well-being, the patient reported a continuously high level in all family members (7 to 9 points on the 10-point scale), whereas her partner reported increasing values in all family members until week 12 (rising from 6 to 9 points) but a decrease at the end of the program (4 points for both children, 6 points for the patient).

For coping, the patient reported a continuously high level (4 to 5 points on the 5-point scale), except for the family rating, which started at 3 points (T1) and improved to 5 points (T2). Formal testing for anxiety, depression, intimacy, cohesion, and satisfaction are shown in supplemental online table 1.

Overall ratings of the program's user-friendliness, as judged by the parents and the adolescent, were good. All participants expressed a desire for more audio and video files.

Discussion

This case report shows that web-based counseling is a feasible alternative for families with parental cancer. First, the participants' monthly feedback indicated that the web-based program helped them to cope. Second, the patient reported improvements in anxiety and in cohesion, flexibility, communication, and satisfaction at the end of the program. Third, both the time spent online and the numbers of pages downloaded were considerable. It is unlikely that more than 20 hours of face-to-face counseling can be provided to a family within four months of diagnosis.

Among the advantages of web-based counseling are that it can be accessed irrespective of time and place, and that participants can spend as much time as needed online and freely choose their topics of interest (9). In H.D.'s family, monthly open feedback was generally positive. However, formal testing – especially in the partner – indicated psycho-social distress at the end of the program, as shown by increased anxiety, and decreased intimacy scores.

Qualitative feedback showed that the adults needed reassurance with respect to the information they gave their children. This was felt to be a major challenge. The support offered by the program was deemed to be important in this regard.

It is interesting to note certain opposed trends in the adults: the patient reported deteriorations in conflict behavior and intimacy after one year, whereas her partner reported improvement. Unfortunately, it is difficult to interpret these results, as these scores were not discussed with the participants. This finding illustrates a disadvantage of online programs following the minimal contact concept; as such findings are not discussed with a psychotherapist. However, test data of this kind could serve as a form of continuous monitoring; a certain level of psycho-social distress could trigger additional support measures, such as face-to-face counseling.

This is the first online cancer counseling program aimed at the entire family, including underage children. Therefore, no direct comparisons can be made. However, results from other web-based interventions for cancer patients (10) show that such interventions can positively influence social and psychological outcomes and health-related quality of life.

Taken together, web-based counseling appears to be a feasible alternative for families with parental cancer. However, until results are available from a larger group of patients and families, no general conclusions about its benefits can be drawn.

Acknowledgments

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Key points

- Protocols for counseling of families with parental cancer are known, but web-based programs for entire families have not been published.
- FAMOCA is a web-based program aiming at the entire family, including minor children.
- The program consists of four modules of four weeks each. All family members have an individual access to the age-adjusted modules.
- This report describes the effects in a family affected by parental cancer immediately after diagnosis, and a follow-up of one year.
- Feasibility is shown by the time spent online and by the subjective effects on coping, anxiety, and family cohesion.

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Table 1: *Formal testing of psycho-social adjustment*

	Patient (HD)			Partner (FD)			Adolescent (ND)		
	T1	**T2**	**T3**	**T1**	**T2**	**T3**	**T1**	**T2**	**T3**
HADS, sum score (0–21)									
HADS-A	16	11	19	9	13	22			
HADS-D	5	8	15	6	7	20			
PFB, sum score (0-30)									
Conflict behavior	1	3	17	19	19	9			
Intimacy	30	28	6	17	14	22			
Mutuality	24	22	14	18	19	16			
FACES-IV, (% of max)									
Cohesion	68	85	84	65	68	58	65	68	68
Flexibility	65	80	80	50	50	35	60	58	58
Communication	74	94	83	18	28	14	86	88	88
Satisfaction	23	71	58	13	12	10	58	40	40

Note: HADS; Hospital Anxiety and Depression Scale; A: Anxiety; D: Depression; a higher score indicating higher tendency for anxiety or depression, PFB; Partnership Questionnaire; higher score indicating higher partnership satisfaction, FACES-IV; Family Adaptability and Cohesion Evaluation Scale

Online Supplemental Appendix (www.famoca.ch)

Table 1: Program content for adults

Written information	How to talk to children about cancer and its significance in everyday life • Importance of a sound diagnosis • How to deal with emotions during information giving • The importance of both partners being present when talking about cancer • How to choose the ideal time talking about cancer • Choosing the ideal environment, an adequate opening, the person to start the conversation, and the content • The importance of the quantity of information • Family hierarchy – which children to inform first • The possibility to include close confidants Improving the maintenance of family functioning, relaxation exercises and promotion of coping strategies Dealing with own emotions and those of other family members; promotion of communication and closeness in partnership Recognizing the family's and children's development; planning the near future as a family; becoming aware of one's own personal and partnership development
Audio	Relaxation exercises, spoken by an experienced psychotherapist using the technique of imagination, each exercise lasting about 20 minutes
Video	Video examples of parent–child conversation showing an example focusing on the ideal environment, the presence of the whole family, a possible opening of the conversation, and dealing with emotions
Online	Contact to other parents in the program (closed group)
Email	Specific questions to be answered by a dedicated psycho-oncologist

Table 2: Program content for adolescents (over 12 years)

Written information	What happens after the diagnosis? • When and how does the therapy start • How long does the therapy last, what possible therapies exist • What happens after the therapy • What does the therapy mean to me and my family Medical knowledge • Methods of cancer work-up • What is the job of different specialists • What are different methods of treatment • What is a side effect, which side effects an occur Stars living with cancer Witnesses explaining their personal experience Frequently asked questions Changes in daily life; own physical discomfort; finding the balance between being present at home and leisure time, autonomy Coping exercises; recognizing one's own development; recognizing changes and development within the family; speaking about wishes and needs
Audio	Relaxation exercise, spoken by an experienced psychotherapist Music to download
Video	Information about cancer and its treatment Videos addressing different kind of emotions and fostering skills for dealing with one's emotions
Online	Contact to other adolescents in the program (closed group) Private diary (blog version)
Email	Specific questions to be answered by a dedicated psycho-oncologist

Table 3: Program content for children (3–12 years)

Written information	Everyday life • Children's story about a family with parental cancer (cancer and its treatment) • Outdoor games • Handicraft and Coloring; instructions for paper animals, mandalas etc. • Diary to download and print out Emotions • Children's story about two families and their dealing with emotions • Handicraft and Coloring; instructions for paper animals, mandalas etc. • Diary to download and print out Coping • Children's story continued (changes in daily life) • Handicraft and Coloring; instructions for paper animals, mandalas etc. • Diary to download and print out Songs and good humor, children's jokes
Audio	Children's stories to listen to Songs to listen to and songs to participate Relaxation exercise, spoken by an experienced psychotherapist
Video	Activities and games for distraction Funny videos
Online	–
Email	–

Table 4: Tests and Abbreviations

Test	Abbreviation	Explanation
Hospital Anxiety and Depression Scale	HADS	Internationally and frequently used; proven reliability and validity; comprehensive 14 item instrument to determine the levels of anxiety and depression[1]
Fragebogen zur Partnerschaftsdiagnostik	PFB	Most commonly used instrument in the German language; good discriminative and prognostic validity and reliability; used to determine the quality of partnership; 31 items that are assigned to three subscales (conflict behavior, intimacy and mutuality); sensitive to changes in the course of couple therapy[2]
Family Adaptability and Cohesion Evaluation Scales IV	FACES IV	Highly cited and commonly used tool; all subscales are reliable and valid. High levels of concurrent, construct, and discriminant validity were shown; commonly used to evaluate the adaptability and cohesion dimensions in family interactions[3]

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Web-based counselling for families with parental cancer: Baseline findings and lessons learned

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Abstract

Purpose: This is the first study in Switzerland to report on psychological adjustment in children of a parent with cancer using a web-based intervention during cancer therapy. *Design/Sample:* 22 families participated in this randomized controlled web-based intervention program. *Methods:* Quality of life and emotional-behavioral well-being of children were examined using child self-reports, and parent proxy-reports. Furthermore, family communication and satisfaction and feedback on the web-based program were assessed. *Findings:* Children's first stage adjustment to parental cancer did not show detrimental patterns. The "lesson learned" in this setting emphasizes the challenge to reach families in need. The web-based program was appreciated as an additional source of information and support in this mostly highly functioning population. *Conclusion:* While feasibility was shown, it remains unclear how to contact families with lower psychosocial functioning.

Keywords: child adjustment, family functioning, lessons learned, oncology, parental cancer, web-based intervention study

Background

In Switzerland, approximately 23,000 men and 19,000 women are newly diagnosed with cancer every year.[1] About 13% of them are diagnosed before the age of 50, a life stage at which having and raising children is normal and common.[2,3] Long-term treatments and the emotional burden of a cancer diagnosis may therefore interfere with parental tasks and responsibilities challenging the entire family system.[4]

Over time, research has broadened its focus on examining the impact of parental cancer not only on the patient, but also on the entire family system.[5,6] Significant levels of distress and mental health problems, including depression, anxiety, and adjustment disorders occur in 32-38% of affected patients and their partners.[7-9] For the children, parental cancer represents a highly stressful situation. Studies have shown that in 25-30% the children of early stage cancer patients show signs of internalizing, externalizing and emotional difficulties including anxiety and mood disorders, psychosomatic problems, aggressive behavior, feelings of guilt and shame, and worsening of academic performance.[10,11] Furthermore, a parental cancer diagnosis comes along with changes in daily routine and role functioning.[10,12] The child`s adjustment to parental cancer is moderated by his or her developmental stage, gender and support network.[13] Adolescent daughters and latency-aged sons report higher levels of psychosocial symptoms than adolescent boys and latency-aged daughters.[10,13-16] In addition, parental psychological functioning, marital satisfaction, parenting skills, family functioning, and communication and coping strategies mediate adjustment to parental cancer, while medical parameters have little impact on the child`s well-being.[10,11,15,17-19]

Taken together, a cancer diagnosis poses high demands on the family system and triggers an adjustment process for the family, the couple and the individual, which may

be challenging especially for families with low resources and pre-existing psychosocial and health problems. For those, psychosocial support and specific interventions are needed to improve functionality of the coping process.[20,21] Inhestern and colleagues summarized several parent-, child-, and a few family-centered interventions and concluded that most of them led to improvements of the quality of life of parents and children, of depression scores and of various aspects of family functioning (e.g. family communication).[5] Literature shows that support offers are often sought out if the parents perceive a need for their children or other family members, and especially if any behavioral changes become apparent in their children.[22,23] Furthermore, Romer and colleagues emphasized that families may make use of counselling following their attending physician's recommendations, because he or she represents a reliable and trustworthy person.[21] This observation is confirmed by the review of Inhestern, et al., showing that the most promising way to reach families is when the support is recommended by healthcare teams.[5]

The reported interventions were realized as face-to-face programs, however in recent years, web-based interventions have started to be assessed more systematically.[24] As cancer patients, relatives, and adolescents commonly use the Internet as a source of information and for support[25], web-based intervention programs have increasingly been shown to be valuable because of the convenience, availability and accessibility of information at all times, as well as for reasons of anonymity.[26] Consequently, web-based interventions seem to be able to overcome some of the known barriers of seeking psycho-oncological support. [5,21]

So far, single, couple and group web-based interventions have been described, while there is still a paucity of scientifically evaluated psychosocial interventions for entire families affected by parental cancer.[3,23,27] We therefore developed and evaluated a

web-based intervention program for families with a parent diagnosed with cancer. The current study 1) reports on self- and parental reported children's quality of life, behavioral-emotional well-being, and family functioning at baseline, and 2) discusses the usability of and the participants' satisfaction with the program as well as the lessons learned from establishing and conducting the study.

Methods

Study design

FAMOCA – *family online counselling for families with parental cancer* was designed as a randomized controlled intervention study. Developed by an interdisciplinary team of adult and child psychologists and oncologists, the study aimed at evaluating the effectiveness of a web-based, interactive and multimedia-based intervention for families with parental cancer. The primary goal was the improvement of child and parental adjustment and family functioning. The intervention group was compared to a control group, which received treatment as usual, consisting of selected, publicly available information brochures on parental cancer for parents and children. The study protocol was approved by the local ethics committee and was registered on drks.de (DRKS00006298).

Participants and procedure

Families were eligible if a parent was diagnosed with cancer for the first time with a high probability of cure, or in the case of metastatic disease with an expected progression-free interval of 12 months. Affected parents had to have at least one child between 3 and 18 years. All family members had to be German speaking and had to have access to the Internet.

Swiss hospitals and cancer centers, physicians and psycho-oncologists were invited to inform patients about the study after disclosing cancer diagnosis. In addition,

families were recruited online by providing study information on cancer-specific websites. Interested families contacted the study team by phone or mail. In a first telephone contact, comprehensive information on the study was given and inclusion criteria were assessed. Written informed consent was obtained individually by each family member. Before randomization (T1) participants completed an online questionnaire, including validated instruments on psychological well-being and familial coping and demographic and cancer specific questions. Five (T2) and 12 months (T3) after study inclusion, the same questionnaires were re-assessed.

Family online counselling intervention

The web-based intervention program was based on the "minimal contact" concept, which implies the participants` autonomous use of the program combined with regular feedback by a professional.[28,29] While participants work individually through educative elements to enhance coping with the current challenges via the web-based program, the therapist contact includes providing a feedback to the participant, responding to questions and unlocking the next module. The program was based on cognitive-behavioral techniques to foster adaption and to build up coping strategies for the entire family.

The online intervention program www.famoca.ch provided individual support for each family member by age-specific platforms for children (3-11 years), adolescents (12-18 years) and adults. While most parts of the different modules were designed for parents to work with their younger children (3-11 years), some elements, such as listening to the story and coloring the pictures were designed for the children to undertake on their own. Every four weeks a new module was activated with the same age-related content for parents, adolescents and children. For example, for children each module included listening to or reading a story of a family with an ill father,

handicraft instructions or a diary to write or draw in. Adolescents watched an educational film clip with cancer information or could use a private blog to process their experience. The content of the modules was adjusted specifically to the first period after diagnosis. A trained psycho-oncologist accompanied every family through completion of the program. The website consisted of four modules:

Module 1: "Understanding what's going on": Families were provided with educational material about communication, coping skills and strategies and information about different cancer treatments and its consequences.

Module 2: "Dealing with everyday family life": This module provided information on the changes in daily life and recommendations on how to maintain family functioning.

Module 3: "Caring for myself and each other": This module assisted families in dealing with emotions aiming to foster parental competences regarding emotional responses in children as well as to enhance emotional interaction within the couple and the family.

Module 4: "Planning the future, integrating the past": This module offered the opportunity to the family to reflect on the changes and the possible progress over the last four months. It helped to integrate new coping behaviors and to identify the resources of each family member to maintain open communication and mutual support.

A detailed description of the program may be found in the publication of Bingisser, et al., 2018.[30]

Study measures

Children`s quality of life (QoL). The QoL of children was measured by the generic KINDL-R questionnaire.[31] It includes self-report forms for children aged 3-6 years and

7-17 years and for each age group a corresponding parent-version. The self-report form for age 3-6 years includes only 12 items resulting in one overall well-being score, which can be compared to parents' reports. It provides good internal consistency (Cronbach's alpha = .80).[31]

*Children`s behavioral-emotional adjustment.* Children`s behavioral-emotional adjustment was measured by the German version of the Strengths and Difficulties Questionnaire (SDQ)[32], a child- and parent reported brief screening questionnaire for children between 2 and 18 years. Adolescents between 12 and 18 years filled in self-ratings. The total score includes results from all subscales except for the prosocial behavior scale. The internal consistency coefficient is 0.82.[32]

Family functioning. Family functioning was measured by the Family Adaptability and Cohesion Evaluation Scale (FACES IV)[33], a self-report questionnaire assessing family satisfaction and communication levels. Parents and adolescents who reached the age of 12 completed this questionnaire. Cronbach`s alpha-Coefficient for FACES IV lies between 0.77 and 0.89.[33]

Use and feedback. Use of the website was examined by analyzing the time spent on the program. After completion of the program, treatment satisfaction was assessed by an evaluation form with open-ended questions, which was developed for this study. Families were asked about the positive/helpful and negative/difficult aspects of the program and responses were categorized. During the study, parents and adolescents responded to a monthly feedback questionnaire, reporting which aspects of FAMOCA were helpful with regard to their own coping and the family's and children's adjustment (5-point Likert scale).

Statistical analysis

The statistical evaluation was carried out using IBM SPSS Statistics 22.0.[34] The recruitment was concluded after the foreseen period of 16 months, but without having reached the target sample size of 90 parent-child dyads. Means, standard deviations and frequencies of the dependent variables at baseline were evaluated separately for groups. In order to verify agreement between the responses of parents and children, the Bland-Altman method was used[35] measuring mean differences (bias) between two measures, with 95% limits of agreement (LoA). The bias was assumed to be significant if the line of equality was not within the confidence interval of the mean difference.[36]

Results

Study sample

A total of 35 direct referrals were made to the study, of which 22 families with a total of 36 children were enrolled. Within these families, four children were not eligible due to their age. Two children and two adolescents declined to participate due to lack of time (n=1) or lack of interest in the content of the program (n=2). In one case the parents did not provide a specific reason for the child's non-participation (n=1). The reasons given by the 13 families who did not participate included: not meeting the inclusion criteria (n=2), lack of need and time (n=4), progression of disease and need of a different kind of support (n=2), and no reason (n=5). In total, 63 participants (34 parents; 29 children) completed baseline measures (table 1). Of the 22 enrolled families, seven were assigned to the control and 15 to the intervention group. Nine families (41%) completed all three measures, three (13%) completed the baseline and the 1-year follow-up measures but left out the post-intervention measures, and ten families (46%) only filled in the baseline measures. There were no significant differences between the families who dropped out and the retained families with regard

to either demographic or medical characteristics (e.g. cancer type). Dropout reasons included a parent's death (n=2), feeling overwhelmed (n=1), lack of time (n=2), unable to reach (n=2), and no reason (n=3). Of the 15 families in the intervention group, seven families (47%) completed all modules, three (20%) completed modules 1 to 3, and five (33%) completed only module 1. Reasons given for completing only module 1 were lack of time (n=3), loss of interest in participation (n=1) and death of a parent (n=1).

Children's quality of life

Children 3 to 6 years. Mean values were compared to norm mean scores of a normal population (table 2).[37] Children had a *total score* within the norm whereas their parents reported lower scores than the norm in the *total score.* The line of equality for the total score was within the confidence intervals of the mean difference. Overall agreement was good across the total score with no points lying outside the 95% limits of agreement. Parents additionally filled in the subscales for their children. They reported lower scores in all subscales compared to the norm.

Children 7 to 17 years. Mean values were compared to norm mean scores of a normal population (table 2).[38] Parents reported slightly lower scores for their children compared to norm values in the *total score* and the subscales *physical well-being*, *emotional well-being*, *well-being in the family* and *well-being related to friends,* and *school-related well-being.* Children showed scores within the norm for all scales except for the subscale *emotional well-being*, in which they scored lower. For all scales the line of equality was within the confidence intervals of the mean difference except for the subscale *well-being in the family*, which may indicate a discrepancy between children's and parents' reports (95% CI Mean of difference; 3.22-16.15). Overall agreement was good across all scales with no points lying outside the 95% limits of agreement.

Children's behavioral-emotional adjustment

Mean values were compared to norm means of a normal population (table 2).[39] Children showed higher means compared to the norm in the subscales *hyperactivity/inattention* and *peer relationship problems*. Parents rated their children between 12 and 18 years higher in the subscale *emotional symptoms*. Parents rated children of all ages higher than the norm in the *total score* and the subscales *emotional symptoms* and *conduct problems*. For all scales, the line of equality was within the confidence intervals of the mean difference. Overall agreement was good across all scales of SDQ ratings with no points lying outside the 95% limits of agreement.

Family functioning

82.4% of parents and 87.5% of adolescents rated communication levels between high and very high. 23.6% of parents and 50% of adolescents rated family satisfaction levels between high and very high while 44.1% of parents and 25% of adolescents rated satisfaction level between low and very low (table 3).

Time spent on the website and feedback

On average, participants spent 32 minutes on the website. Parental feedback was given by phone after completing the program. The content of the website corresponded to the experiences of parents, reassuring them that their diagnosis-related reactions towards their children were adequate. In general, families appreciated the variety of provided information and the freedom to work independently with the program and the monthly phone contact with the psychologist. Children and adolescents liked the active parts of the program (e.g. storybook). Most families criticized that FAMOCA provided too much text to read; some patients mentioned concentration problems due to cancer treatment and children were challenged with their daily schedule. Table 4 provides the categorized feedbacks.

Parents' monthly feedback about the helpfulness of the FAMOCA program for themselves, for the family and for the children showed average scores between 2.6 and 3.3 (n=12). Adolescents' monthly rating showed average scores between 3 and 3.6 (n=3).

Discussion

A primary aim of this randomized controlled web-based intervention study was to describe children's well-being and quality of life as well as family functioning shortly after a parent's cancer diagnosis by comparing the perspective of children and parents, respectively. Additionally, feedback on the benefits and drawbacks of working with the program was evaluated.

Our main results suggest an oligo-symptomatic adjustment process to the parental cancer diagnosis with children and adolescents showing no clinically relevant levels of behavioral and emotional problems and normal levels of QoL. This is congruent with former research reporting that approximately 70% of the children with a parent who has cancer do not show increased levels of substantial psychosocial problems, while 30% of them respond more severely to the diagnosis.[10,14,19] These findings in children are comparable to the number of adults with severe distress symptoms compared to those with transient distress symptoms.[7,9] Increased levels of distress are mostly associated with adverse family and parenting variables, such as poor family functioning, maternal depression, or low parental QoL.[13-15,18] In our study, family functioning with regard to family communication was rated as high to very high by the majority of parents and adolescents. On the other hand, the ratings for level of family satisfaction, a measure for the degree of how happy and fulfilled family members feel with each other, were low in both groups. The disclosure of a cancer diagnosis may lead to concerns and destabilization of the family system which can cause lower levels

of family satisfaction. However, the premorbid degree of satisfaction and happiness in these families is unknown. Child self-reports on QoL and behavioral-emotional adjustment did only differ in family related QoL from parent proxy reports, which may imply an underestimation of QoL in this dimension when judged by parents as compared to their children. Parents tended to report lower scores as compared to their children in overall measurements of QoL and behavioral-emotional adjustment. Previous findings showed that parents reported fewer behavioral and emotional problems and lower QoL scores in their children than the children reported.[10,14,40]

As this is the first study in Switzerland to evaluate an online tool in a family setting, it was especially important to analyze and understand the benefits as well as the drawbacks of the participants using the program. In general, FAMOCA was appreciated and its influence on the adjustment process was rated as moderate. Families felt taken care of and reported that their specific questions and concerns were addressed. Having support in how to communicate with each other seemed to be an important issue. The interactive parts, with relaxation exercises and handicraft instructions were perceived as useful. The children storybook was generally appreciated. However, the resources of the parents and children to work on the program were limited. Family time was restricted due to cancer treatment and daily tasks and-, consequently, activities unrelated to the parent's cancer were often preferred to working on the program. Adolescents preferred other sources of information and for some parents the program did not add to what they knew already. Accordingly, the program was especially helpful for families with a newly diagnosed parent. In these cases what was needed was low-threshold support and specific information on cancer and family rather than intensive psycho-oncological care.

Limitations

The current study has some limitations. Generalizability is limited due to a possible selection bias. Our sample mainly consisted of families with no migration background, good general resources and a high income and education level. The response to cancer is influenced by the cultural background of the patient and his or her family. Most of the couples were married and in most cases the mother suffered from breast cancer. Therefore, the informative value is limited to the described study population. Besides, the program was only available in German.

The small sample size was a key limitation and precluded studying intervention effects. Recruitment turned out to be one of the major challenges. Assuming that the population studied was familiar with the Internet, study information was posted on cancer specific websites focusing on psychosocial issues. We expected that affected parents would contact the study team more often as the FAMOCA website appeared within the top five to ten positions when searching for family, cancer, and support on German google websites. In our study face-to-face recruitment was more powerful than passive online recruitment. Of 22 participating families only three signed in for the study over the Internet, whereas 14 were recruited by their attending physician or psycho-oncologist. However, the number of patients directly referred remained low. Physicians' workload, the predominance of purely medical compared to psychosocial issues may count for the low number of families recruited by oncology staff. Accordingly, a significant personal and financial investment is necessary to create a campaign with a certain impact in which patients are directed to and motivated for an online-study by a trustworthy healthcare team. Therefore, healthcare teams need to be trained to identify families at risk.

Attrition from the study was high: 54% of the participants completed post-treatment measures and 41% follow-up measures. One frequently mentioned dropout reason given by children and adolescents was lack of time or loss of interest. Baseline data of children and adolescents were mostly within the normal range, which indicates a good adjustment to the parental cancer diagnosis. After being initially motivated and curious children and adolescent might later have lost momentum due to their daily routines. In line with previous research on what families need, the content of the program was created according to the presumed concerns arising immediately after disclosure of diagnosis, such as how to talk about cancer within the family. However, nearly one third of participating families completed module one only and then dropped out. One reason could be that families thought themselves as being beyond the point where additional information and exercises on family and partnership communication could be beneficial. Not fully met expectations of participants have been suggested to be one reason for low engagement in web-based interventions.[41] Reflecting the demands of a long-term adjustment process after a parental cancer diagnosis, the modules focusing on dealing with emotions and enhancing coping strategies were found to be helpful by the remaining sample. Because FAMOCA contains various components more research is needed to define in detail which components are more beneficial than others.

The enrollment rate was poor, with 22 participating families compared to the 90 required to achieve an 80% participation rate. One possible reason could be that the burden of affected families to deal with a cancer diagnosis on the one hand and to maintain the daily routines on the other hand is so heavy that participating in a study may at first seem more like an additional strain than a relief. This corresponds to the fact that the majority of the participating families were already in the process of

receiving cancer treatment when a first adjustment process had already been made. In addition, interest in participating in support interventions seems to be low in newly diagnosed patients.[21]

Conclusion

To our knowledge, this study is the first in Switzerland evaluating a web-based intervention program for the entire family affected by parental cancer. Due to the small sample size evidence about the feasibility of the FAMOCA program is limited. Baseline data of children and family parameters revealed a good spontaneous adjustment to the cancer diagnosis, which may have influenced attrition rate. Future studies need to investigate on how to better address the population in need – families with psychosocial cancer-related difficulties, and families with a higher pre-existing psychosocial burden and lower income, respectively. As support offers will be more readily accepted when recommended by a healthcare team, referrals to counselling may be more successful if made by the attending physician. Future studies should invest in the development of appropriate screening instruments and training of medical staff in the detection of distressed families. Additionally, future studies should focus on identifying the subgroups of families in need of psychological support and to implement this knowledge into the recruitment procedure. According to the participants' feedback concerning improvement of the website, all modules of FAMOCA should be made accessible simultaneously. Families may then decide freely which of the chapters of the different modules (e.g. communicating with adolescents) would apply to them. Furthermore, the text should be shortened, because, participant's motivation to follow short and concise recommendations was higher compared to having to extract the important pieces of information from a written text without these specific instructions. Almost all families appreciated recurrent monthly telephone contact. Therefore, it

would be interesting to filter out families likely to benefit rather from a direct therapist contact than from the web-based format. In general, further research is needed to develop made-to-measure cancer-related interventions for the children, the parents and the whole family.

Implications for Psychosocial Oncology

- This first web-based program for the entire family including young children affected by parental cancer showed favorable adjustment to the parental cancer diagnosis in the majority of participants. Parental assessment of the children's well-being tended to be lower than children's self-reported assessment.
- Low recruitment rates and high dropout rates may hamper the use of such programs, particularly for children. Therefore, face-to-face counselling and step-up programs may not be fully replaceable by web-based programs. However, such programs are feasible and the majority of participants report on substantial support.
- The focus of such programs should be on providing cancer-specific information, on dealing with emotions, and on enhancing coping strategies.

Conflict of interest

The authors declare no conflict of interest.

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Table 1. Sample characteristics of participating families

Parents characteristics	n	%
Ill parents	20	
Mothers	19	95
Fathers	1	5
Age in years: Mean=43.2, range=32-51		
Spouses	14	
Mothers	2	14.3
Fathers	12	85.7
Age in years: Mean=43.8, range=34-55		
Highest level of education completed by ill parents		
Low	0	
Middle	10	50
High	10	50
Highest level of education completed by spouses		
Low	0	0
Middle	4	28.6
High	10	71.4
Children and adolescents characteristics	**n**	**%**
Children	20	
Daughters	10	50
Sons	10	50
Age in years: Mean=8.2, range=4-11		
Adolescents	8	
Daughters	2	25
Sons	6	75
Age in years: Mean=13.4, range=12-17		
Family characteristics	**n**	**%**
Single-parent families	5	15.2
Number of children in a family		
1	8	36.4
2	11	50
3	3	13.6
Number of children participating in the study per family		
1	11	57.9
2	7	36.8
3	1	5.3

Table 2. Means, standard deviations and Bland-Altman limits of agreement of children's quality of life and behavioral-emotional well-being.

	Baseline								
	Norm-values	Parents	Parents [a]	Norm-values	Children	Bland- Altman LoA	$M_{difference}$	$SD_{difference}$	CI of $M_{difference}$
		M (SD)	M (SD)		M (SD)				
KINDL-R (3-6 years)		n = 7			n = 7				
Total score	80.0	78.00 (7.28)	73.43 (1.90)	73.0	73.43 (1.90)	-19.07; 9.93	-4.57	7.44	-11.45; 2.31
		n = 14							
Physical well-being	80.2	74.55 (17.24)	-		-	-	-	-	-
Emotional well-being	83.0	79.76 (14.04)	-		-	-	-	-	-
Self-worth	73.6	72.32 (15.25)	-		-	-	-	-	-
Well-being in the family	80.7	75.00 (21.78)	-		-	-	-	-	-
Well-being related to friends	79.7	74.55 (12.85)	-		-	-	-	-	-
School-related well-being	83.8	78.57 (13.59)	-		-	-	-	-	-
KINDL-R (7-17 years)		n = 37	n = 22		n = 22				
Total score	76.9	75.87 (11.54)	75.83 (10.19)	73.0	77.70 (7.33)	-17.95; 14.22	1.87	8.25	-1.79; 5.53
Physical well-being	77.2	76.01 (20.28)	74.66 (18.89)	70.7	76.99 (14.35)	-39.58; 34.92	2.33	19.10	-6.14; 10.78
Emotional well-being	80.9	77.03 (14.66)	77.01 (13.54)	81.6	76.42 (11.07)	-25.82; 27.00	-0.59	13.54	-6.59; 5.41
Self-worth	69.7	74.49 (13.21)	75.22 (11.14)	58.4	72.16 (15.16)	-28.26; 34.38	-3.06	16.06	-10.18; 4.06
Well-being in the family	78.3	74.83 (15.34)	74.41 (13.06)	82.5	84.09 (10.16)	-38.12; 18.76	9.68	14.58	3.22; 16.15
Well-being related to friends	78.0	77.70 (13.38)	77.33 (12.50)	77.5	79.83 (8.82)	-25.82; 20.82	2.50	11.96	-2.80; 7.80
School-related well-being	77.0	75.17 (19.40)	75.86 (17.51)	67.2	76.70 (20.23)	-27.19; 25.51	0.84	13.52	-5.15; 6.83
SDQ	Norm-values	Parents M (SD) n = 59	Parents [b] M (SD) n = 8	Norm-values	Children between 12 and 18 years M (SD) n = 8	Bland- Altman LoA	$M_{difference}$	$SD_{difference}$	CI of $M_{difference}$
Total score	8.4	9.61 (4.99)	8.63 (4.10)	10.3	10.63 (3.07)	-5.66; 9.66	2.00	3.93	-1.29; 5.28
Emotional symptoms	1.9	2.53 (2.18)	2.38 (1.96)	2.8	2.88 (2.42)	-3.54; 4.54	0.50	2.07	-1.23; 2.23
Conduct problems	1.6	1.92 (1.52)	1.38 (1.06)	2.2	1.25 (0.89)	-2.32; 2.07	-0.13	1.13	-1.07; 0.82
Hyperactivity/Inattention	3.5	3.56 (2.58)	3.50 (2.62)	3.8	4.00 (1.07)	-3.67; 4.67	0.50	2.14	-1.29; 2.29
Peer relationship problems	1.5	1.61 (1.59)	1.62 (1.51)	1.5	2.50 (1.51)	-2.49; 4.24	0.88	1.73	-0.57; 2.32
Prosocial behavior	8.6	9.61 (4.99)	8.25 (1.04)	8.0	8.38 (1.19)	-3.24; 3.49	0.13	1.73	-1.32; 1.57

Note: [a] Parents proxy reports of children who filled out self-reports; [b] Parents proxy reports of children between 12 and 18 years; M, Mean; SD, Standard deviation; LoA, 95 percent limits of agreement; $M_{difference}$, mean difference; CI, confidence interval; SDQ, Strengths and Difficulties Questionnaire. Higher scores in the KINDL-R reflect better ratings; higher scores in the SDQ reflect greater problems, except for the prosocial behavior subscale.

Table 3. Frequencies, means and standard deviations of communication and satisfaction levels of parents and adolescents (12-18 years).

FACES-IV		Baseline			
		Parents (n = 34)		Adolescents (n = 8)	
		n (%)	M (SD)	n (%)	M (SD)
Communication level	Very low	1 (2.9)	73.47 (22.27)	0	80.25 (10.34)
	Low	2 (5.9)		0	
	Moderate	3 (8.8)		1 (12.5)	
	High	14 (41.2)		3 (37.5)	
	Very high	14 (41.2)		4 (50.0)	
Satisfaction level	Very low	5 (14.7)	45.18 (24.86)	1 (12.5)	62.25 (27.71)
	Low	10 (29.4)		1 (12.5)	
	Moderate	11 (32.4)		2 (25.0)	
	High	5 (14.7)		2 (25.0)	
	Very high	3 (8.8)		2 (25.0)	

Note: M, Mean; SD, Standard deviation; FACES-IV, Family adaptability and cohesion evaluation scale. FACES-IV levels can vary from ‘very low’ to ‘very high’.

Table 4. Feedback of families on the FAMOCA program

Positive Feedback on the FAMOCA program (What did families like the most? What was helpful?)	
Parents	• Context of the program corresponded to own experiences, which strengthened the belief that their responses and actions were adequate (n = 11) *"I felt reassured that we informed our children early enough"* • Variety of provided information (n = 8) • Working independently with the program (n = 3) *"We liked that we did not have to find one schedule for all family members together"* • Recommendations on how to talk with the children and each other (n = 4) *"Good to receive support talking with the children and knowing how important it is, since they feel the change anyway"* • Working as a family together on the program (n = 2) *"It was nice to spend time with my husband working together on FAMOCA"* • Monthly phone contact with the psychologist (n = 6) • Specific questions and concerns were addressed during the program (n = 5) • Children asked more questions (n = 2) • Relaxation exercises (n = 9)
Adolescents	• Examples of songs (n = 1) *"Downloading the songs was better than reading"* • FAMOCA helped finding specific information (n = 5)
Children	• Storybook (n = 8) *"My children could hardly wait for the children's story book"* • Handicraft instructions (n = 7) *"We loved the artwork of the sorrow doll"* • Downloading songs (n = 3) • Writing in the diary (n = 5)
Negative feedback on the FAMOCA program (What was difficult?)	
Parents	• Too much text (n = 8) *"Since my concentration was low, the text was too long for me to read"* • No new content, only helped shortly after diagnosis (n = 5) *"Some information came too late because I am already through with the chemo"* • Too many and difficult questionnaires (n = 4) • Exhausting to accompany the children through the different modules (n = 2) • Lack of time (n = 4) *"Family time is already limited. We did not want to spend it in front of the computer"* • Too little information for single parents (n = 1)
Adolescents	• Too much text (n = 2) • No time and no interest in the content of the program (n = 2) • No need, found information elsewhere (n = 4) • Too much confrontation with cancer (n = 4)
Children	• Too much to read (n = 5) • Worksheets remind of school (n = 1) • Children have enough challenges from school (n = 2) • Little time or lack of motivation due to a full daily schedule (n = 5)

Feasibility of minimal contact interventions is limited in couples affected by a new cancer diagnosis

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Abstract

Objective: Patients with recently diagnosed cancer show high rates of distress and may develop depression and anxiety. Their partners are also affected by psychosocial stress, but interventions aiming at couples remain to be a challenge. Therefore, this study examined the feasibility of minimal contact interventions and psychological adjustments in couples immediately after a cancer diagnosis.

Methods: Observational study on minimal contact interventions using psychometric testing in patients and their partners before and 16 weeks after initial therapy, typically surgery, followed by chemotherapy. Depression, anxiety, optimism, pessimism, and quality of relationship were assessed. Written information conveyed to all eligible patients, a specifically designed homepage, and active personal information of all oncologists in the University Hospital Basel were used for recruitment. Feasibility was defined as an inclusion rate over 50% in eligible couples and a dropout rate under 50% in couples included.

Results: 292 couples were eligible, 33 couples showed interest and were screened, and 20 patients and 14 partners could be included. 17 patients underwent surgery and chemotherapy. Inclusion rate was 61% in patients screened and 7% in patients eligible. Dropout in patients at 16 weeks was 50%. Anxiety was pronounced at diagnosis in patients and partners, but decreased during follow-up. Optimism was subdued at diagnosis, but increased during follow-up.

Conclusion: Feasibility of minimal contact interventions was limited in our sample, if eligible patients were to be considered. However, the majority of effectively screened patients could be included, half of them showing high adherence. It remains unclear, if improvements in optimism and anxiety can be attributed to the minimal contact interventions. We conclude that minimal contact interventions may be used for the support of couples affected by parental cancer, but the majority of patients and partners do not actively reach out to use such programs.

Objectives/Introduction

Cancer not only affects patients, but also nearest relatives and particularly partners (2004; Hagedoorn, Sanderman, Bolks, Tuinstra, & Coyne, 2008). A new cancer diagnosis confronts patients and those close to them with an enormous amount of

psychosocial stress (McClure, Nezu, Nezu, O'Hea, & McMahon, 2012), challenging psychosocial adjustment, communication, coping, and social support of those directly and indirectly affected (Brandao et al.). Rates of depression and anxiety are significantly higher in cancer patients than in the general population with point prevalence estimates of about 20% (Linden, Vodermaier, Mackenzie, & Greig, 2012; Mitchell et al., 2011). Similar findings were found in partners of patients with cancer (Mitchell et al., 2011). While overt depression and anxiety was described in 10% of partners, subclinical signs were found in up to 30%. Considering this, several couple-based face-to-face interventions to improve the patients' and partners' quality of life have recently been published (Li & Loke, 2014). However, due to a certain lack of knowledge on spousal communication (Badr, 2017) and limited resources, effective psychosocial interventions are not conveyed to all families in need – even in high-income countries, such as Switzerland, where over 40% of patients undergo in-hospital rehabilitation (Ture et al., 2015).

Here, minimal contact interventions, such as programs based on written information or information provided by the Internet, could possibly serve as tools to support patients and partners with newly diagnosed cancer. The strengths of these interventions are the low cost, the high standardization, and particularly the independence of time and place. This may be specifically important to partners who mostly remain in their social and work-related routine. However, disadvantages of minimal contact interventions are missing personal contact to psycho-oncologists, as well as a pronounced dependency on information technology, health literacy, and motivation to use the provided information independently. Minimal contact interventions have proven effects in many different fields, such as anxiety (Axelsson, Andersson, Ljotsson, & Hedman-Lagerlof, 2018), irritable bowel syndrome (Pajak, Lackner, & Kamboj, 2013), and depression (Newman, Szkodny, Llera, & Przeworski, 2011). Unfortunately, there are only limited results regarding feasibility and efficacy in such interventions focusing on patients (Urech et al., 2018) or families (Bingisser et al., 2018) in newly diagnosed cancer. One of the problems reported with minimal contact interventions was the low inclusion and the high dropout rate (Badr & Krebs, 2013; Zimmermann, 2015) or generally speaking the feasibility of such programs. We therefore report on the feasibility, as defined by inclusion rate and adherence, of minimal contact interventions in patients and their partners with underage children during the first 16 weeks of cancer therapy in a Swiss University hospital. Specific aims were to study use and adherence to the programs

and report on psychological adjustment in patients and partners at inclusion and after 16 weeks on the minimal contact intervention.

Methods

We collected data as a part of the randomized controlled web-based intervention study "FAMOCA – family online counselling for families with parental cancer" aiming to improve psychological adjustment in families of newly diagnosed cancer patients. This program was developed based on cognitive-behavioural therapy interventions by focusing on psycho-educative elements, enhancing open communication, affective involvement, family and relationship cohesion, problem-solving skills, and adaptive coping at the level of the individual, the couple, and the family. The detailed description of the program can be found in a recent publication (Bingisser et al., 2018). Informed consent was obtained from all individual participants (patients and partners). Ethic approval was obtained from the responsible local ethics board (EKNZ 38/13).

Participants and inclusion criteria

Eligible patients had to present with a newly diagnosed cancer (within last month), an expected progression free survival of 12 month, at least one child between 3 and 18 years, and with sufficient German speaking and reading skills.

Procedures

Patients were recruited by provision of leaflets in their information folder at hospitalization, in waiting rooms, and online forums between March 1st 2013 and June 30th 2015. Families were motivated to contact the research team by oncologists, through the website, by email or by telephone. Participants were provided with detailed information and terms of participation. A written informed consent form was individually obtained from all participants. After screening of all patients showing active signs of interest through an email contact or a phone call, participants were assigned either to an internet-based intervention or a comprehensive self-administered written information, based on contents provided by the Swiss and German Cancer Leagues. Patients and their partners had regular email contact with the study team in both groups, and monthly assessments on the use of support, treatment satisfaction, coping, and individual wellbeing.

T1 was defined as the start of the program, and T2 was defined as the completion of the intervention, 16 weeks after T1. Participants filled in identical questionnaires (see below) at T1 and T2. The number of eligible patients with newly diagnosed cancer and underage children were retrieved from the hospital's electronic health records (EHR). Reasons for non-enrolment of screened patients were recorded; they were categorized into failure to meet inclusion criteria, (e.g. inadequate computer skills, palliative treatment, lack of family member availability), lack of time (e.g. due to family schedules), and unwillingness to participate.

Measures

Demographic and cancer specific data were gathered from self-reports and medical reports using the EHR. Demographic data included age, gender, marital status, number of children, educational level, monthly income, cancer diagnosis, and type of treatment (see table 1). In both groups, questionnaires were to be completed online at T1 and T2, and included the following tools:

Depression and Anxiety: To assess the patients' and partners' anxiety and depression, the German version of the Hospital Anxiety and Depression scale HADS was utilized (Herrmann-Lingen, Buss, & Snaith, 2011). The HADS is a self-report questionnaire, which has been specifically developed for physically ill patients. It consists of 14 items and is divided into an *Anxiety* subscale (*HADS-A*) and a *Depression* subscale (*HADS-D*), both containing 7 items. The items are scored on a 4-point Likert-scale ranging from 0 to 3. Cronbach's alpha across different studies varies between α = .68 and α = .93 (mean α= 0.83) for *HADS-A* - and between α = .67 and α = .90 (mean α = .82) for *HADS-D*. Cut-offs were defined as $\geq$9 (9/21) for *HADS-A,* and $\geq$8 (8/21) for *HADS-D,* because of the equilibrium of sensitivity (.80) and specificity (.80).

Optimism and pessimism: The German Version (Herzberg, Glaesmer, & Hoyer, 2006) of the Life Orientation Test (LOT-R) is a revised version of the original LOT (Scheier, Carver, & Bridges, 1994) a 10-item measure of optimism versus pessimism. Of the 10 items, 3 items measure optimism (LOT-O), 3 items measure pessimism (LOT-P), and 4 items serve as controls. Respondents rate each item on a 4-point scale: 0 = *strongly disagree,* 1 = *disagree, 2* = *neutral,* 3 = *agree,* and 4 = *strongly agree.*

Quality of marital relationship: The self-reports of quality of marital relationship were evaluated with a marital relationship questionnaire *Partnerschaftsfragebogen* PFB (Hahlweg, 1996). The questionnaire contains three subscales, *conflict behaviour*, *tenderness,* and *communication*. Each subscale consists of 10 items, which is scored on a 4-point Likert scale ranging from 0-3. Sum scores range from 0-90 and high sum scores indicate a high quality of marital relationship. Cronbach's alpha for the three subscales are α = .88 for *conflict behaviour*, α = .91 for *tenderness*, and α = .85 for *communication*. The following cut-offs were used: *conflict behaviour* 5.4; *tenderness*: 20.1; *communication*: 20.1; *total sum score*: 64.9 (16).

Coping styles: The German Version of the Brief COPE (Carver, 1997) consists of 28 Likert-scaled items and assesses 14 coping styles: self-distraction, active coping, denial, substance use, use of emotional support, use of instrumental support, behavioural disengagement, venting, positive reframing, planning, humour, acceptance, religion, and self-blame. Each item was scored on a 4-point scale; each category contained two questions of maximum 4 points each. The number of points are an indication for the style of coping, the maximum per category being 8 points.

Statistical analyses

The statistical evaluation was carried out using IBM SPSS Statistics 22.0 (IBM Corp.). Means, medians, and distributions were calculated separately for all groups and time points. For comparisons, two-tailed Wilcoxon signed-rank or T-tests were used for dependent samples, and Levene tests for independent samples, where appropriate.

Results

292 patients were theoretically eligible between December 1st 2013 and June 30th 2015 according to the hospital's database. 33 patients actively contacted the study team, and 13 patients could not be included due to palliative treatment in two patients, lack of family member availability in two patients (either partner or children unwilling or unable to participate), restricted family schedule (school, sports) in four patients, and unwillingness to sign informed consent for no specific reason (see figure 1). Also, one couple was formally included, but had to be excluded due to unexpected early death.

In sum, 20 patients consented to the study resulting in an enrolment rate of 61% in screened patients and 7% in eligible patients, respectively.

Seven couples (13 patients, 9 partners) were assigned to the internet-based intervention and five couples (7 patients, 5 partners) were assigned to the self-administered information intervention. In order to assess the feasibility and effects of minimal contact interventions, patients and partners were pooled, respectively. There were no significant differences in demographic or outcome variables between the groups, as well as between participants completing the program and dropouts at T2. Demographics are shown in table 1. For comparisons between T1 and T2, dropouts were excluded. Patients included suffered from the following cancers: 13 had breast cancer, 2 had lung cancer, and 5 had gallbladder, cervix, appendix, pancreatic, and skin cancer, which were treated with 17 chemotherapies, 17 surgeries, 12 radiotherapies, and 12 other therapies.

Depression and Anxiety: HADS-D for depression showed sub-threshold (<8) scores in the majority of patients (median 6) and partners (median 6.5) at T1, without significant change at T2 ($t=1.38$, $p=0.20$). HADS-A for anxiety levels were above the cut-off (≥ 9) scores in the majority of patients (median 11) and partners (median 10), with significant decrease in patients at T2 ($t=3.23$, $p=0.01$) (see table 2).

Optimism and pessimism: LOT-P showed mildly elevated pessimism scores for the majority of patients (median 4.5 of 12) and partners (median 3 of 12) at T1, without significant change at T2 ($t=-0.12$, $p=0.91$). LOT-O showed subdued optimism scores for the majority of patients (median 5 of 12) and partners (median 6 of 12), without significant increase in patients at T2 (see table 2).

Quality of marital relationship: PFB showed intermediate scores for the majority of patients (median 69.5 of 90) and partners (median 52.5 of 90) at T1, without significant change at T2 ($t=1.27$, $p=0.22$). Sub-scores were comparable between patients and partners; the only exception being a significant difference in the rating of conflict behaviour between patients (median 5) and partners (median 9) at T1 ($t=-2.17$, $p=0.04$). This difference was consistent at T2 (see table 2).

Coping styles: The BCOPE at T1 and T2 showed high scores (6 to 8 points) for the following coping styles in patients: active coping, use of emotional support,

instrumental support, positive reframing, planning, and acceptance. Ratings of partner and patient's BCOPE measures at T1 and T2 showed low scores (2 to 4 points) for the following coping styles: denial, substance use, behavioural disengagement, humour, religion, and self-blame (see table 3).

Discussion

The main finding of our study was the limited feasibility of minimal contact interventions in newly diagnosed cancer, if the inclusion rate of theoretically eligible patients was taken as the definition. However, the majority of effectively screened patients were willing to participate, and full adherence to the interventions was shown in 50% of all patients. Possible reasons for the low participation of eligible patients were the form of recruitment via oncologists and written information at hospitalisation, the timing around the start of cancer therapy, and the competition with face-to-face counselling. The majority of withdrawals at screening were due to time issues and lack of interest. This finding supports the experience of many caregivers at the start of cancer therapy, particularly in patients with underage children: The most often female patients are overwhelmed by the dramatically new situation, dominated by healthcare appointments and simultaneous organisation of occupation, family and other social chores. It must be pointed out that patients with children are in an age group demanding to the entire family regarding career, child raising, finances, and social integration. Particularly women tend to take over many different roles, and the raising of the children is in their utmost interest – a steady state has often not been reached. Therefore, newly diagnosed cancer may severely hit an entire family; an already complex schedule is completely thrown over. All these problems may explain difficulties in inclusion and adherence in families affected by parental cancer.

Patients with cancer generally have a low rate of psychosocial conditions at diagnosis, in spite of a high distress, but may develop depression and anxiety disorder in the course of disease (Henselmans et al., 2010). In our patients, there was a high level of anxiety at inclusion, with a decrease during the first 16 weeks of therapy. Optimism was restrained at the start of therapy, but increased at follow-up. Interestingly, our patients reported several positive coping styles - with markedly high scores – such as active coping, the use of emotional support, the use of instrumental support, positive

reframing, planning, and acceptance. These strategies have been shown to be associated with quality of life in cancer patients (Shapiro, McCue, Heyman, Dey, & Haller, 2010). Similarly, the BCOPE showed low scores for negative coping styles such as denial, substance use, behavioural disengagement, and self-blame. Taken together, the participating patients and partners seemed highly selected for positive behavioural traits. Furthermore, monthly feedbacks with psycho-oncologists showed high emotional and communicative skills in almost all patients included.

How do these results compare to the literature? First, there is very little evidence for minimal contact interventions, and almost none in families with newly diagnosed cancer. A recent study (Urech et al., 2018) showed that quality of life was significantly higher and distress significantly lower in an intervention group using a web-based minimal contact program, as compared to a group waiting for intervention. However, changes in anxiety or depression were not significant in the intention-to-treat population. Further, recruitment was more actively carried out, and an inclusion rate of over 50% was reported. However, this study was performed in patients only, irrespective of family participation, and the inclusion period was extended to three months after the start of first-line therapy as compared to one month after diagnosis in the present study. One might conclude that three months are more suitable in such interventions, as the first month is a specifically vulnerable period. However, effects of an intervention may be different and even less needed three months after the start of therapy. Comparable minimal contact interventions have so far only shown general acceptability (Karageorge et al., 2017), they have been restricted to physical activity behaviour change (Forbes, Blanchard, Mummery, & Courneya, 2017), or they have focused on the role of the therapeutic facilitator (Carter, Fergus, Ahmad, McLeod, & Stephen, 2015). In our opinion, minimal contact interventions may have a place in the future, particularly in the early phase of cancer treatment in individual patients, but possibly also in their partners. Unfortunately, the benefit of psychosocial interventions for couples coping remains unclear (Zimmermann, 2015), partly due to recruitment problems. A recent meta-analysis showed that almost half of all interventions recruited less than 35 couples per group, refusal rates reaching 82% (Badr & Krebs, 2013). To be prudent, one might conclude that the novel tool of minimal contact intervention is yet another possibility to support families affected by parental cancer. Reasons for low recruitment and considerable dropout cannot be finally judged, the necessity to use

computer and Internet being one of the possible reasons. On the other hand, the present results show similar dropout rates in patients and partners provided with written information, which puts the Internet as a deterring mechanism into perspective. Certainly, increasing habituation to Internet technology will support the use of minimal contact interventions in the future. Nevertheless, more studies are needed in search of the reasons of low inclusion and high dropout in minimal contact interventions in order to facilitate the use of such cost-effective and highly standardised tools.

Limitations

This study has multiple limitations. First, the number of participants was low. Therefore, a differential assessment of outcomes regarding the two different minimal contact interventions was not possible. Second, most patients included were women with breast cancer. They tend to undergo most of their therapy in an ambulatory setting. According to a recent publication in Switzerland (Ture et al., 2015) they do not use in-patient rehabilitation facilities very often, which is clearly different in other types of cancer. Therefore, the results should not be generalized. Third, participants were patients in 60% and affected partners in 40%. Individually differing reactions to interventions are well known – therefore, the spread of clinical effects may be even wider in a mixed group of patients and partners. Interestingly, most psychometric assessments were comparable between patients and partners, except for conflict behaviour, where partners seem more sensitive, while patients might be preoccupied with disease and treatment. Fourth, in spite of one of the big advantages, namely the standardisation of information given using such minimal contact interventions, there is no good evidence on the content of such information (Badr, 2017). Therefore, direct comparison to face-to-face counselling should be attempted, where the individualised contact focusing on the therapeutic relationship could turn out to be an advantage of direct counselling.

Conclusion

Minimal contact interventions, such as an online-program or a comprehensive written information program – in spite of good evidence in other fields – are yet at an early stage in newly diagnosed cancer, particularly if focusing on patients and their partners together. Feasibility may be hampered by the higher effort patients and partners need to take, by the use of information technology, and by the lack of face-to-face counselling, which remains to be the golden standard in patients, partners and children affected by newly diagnosed parental cancer.

Conflict of interest

The authors declare no conflict of interest.

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Table 1. Baseline characteristics

		Patients (n = 20)	***Partners (n = 14)***
Gender			
	Male	1	12
	Female	19	2
Mean age *(years)*		43.2	43.8
(SD/range)		(5.6; 32 – 51)	(5.7; 34 – 55)
Marital status			
	Married	16	13
	Unmarried	3	0
	Divorced	1	1
Children (n)			
	1	7	7
	2	10	5
	3	3	2
Educational Level			
	Low	0	0
	Middle	10	4
	High	10	10

Table 2. Psychometric testing

	T1 Patients M (range)	T2 Patients M (range)	T1 Partner M (range)	T2 Partner M (range)
HADS	n = 20	n = 10	n = 14	n = 6
Depression	6.00 (13)	5.50 (10)	6.50 (14)	7.50 (16)
Anxiety	11.00 (10)	8.00 (13)	10.00 (10)	6.50 (11)
LOT-R	n = 20	n = 10	n = 14	n = 6
Pessimism	4.50 (15)	3.00 (6)	3.00 (15)	9.50 (11)
Optimism	5.00 (6)	6.00 (8)	6.00 (4)	5.50 (5)
PFB	n = 16	n = 9	n = 12	n = 5
Conflict behaviour	5.00 (11)	5.00 (10)	9.00 (22)	12.00 (17)
Tenderness	20.00 (25)	19.00 (26)	17.00 (26)	13.00 (14)
Communication	22.50 (20)	20.00 (14)	19.50 (22)	19.00 (13)
Total score	69.50 (46)	64.00 (45)	52.50 (55)	45.00 (39)

Note: M: median; n: number of participants; HADS: Hospital Anxiety and Depression Scale; LOT-R: Life Orientation Test; PFB: Partnerschaftsfragebogen (quality of marital relationship)

Table 3. Coping styles

	T1 Patients M (range)	T2 Patients M (range)	T1 Partner M (range)	T2 Partner M (range)
BCOPE	n = 20	n = 10	n = 14	n = 6
Self-distraction	5.50 (4)	4.50 (4)	5.00 (4)	4.50 (4)
Active coping	8.00 (4)	6.00 (6)	5.00 (5)	5.00 (4)
Denial	3.50 (5)	3.00 (3)	3.00 (5)	2.00 (3)
Substance use	2.00 (3)	2.00 (0)	2.00 (5)	2.00 (4)
Use of emotional support	8.00 (5)	7.50 (4)	6.50 (4)	6.00 (3)
Instrumental support	6.00 (5)	6.50 (6)	5.50 (6)	4.50 (6)
Behavioural disengagement	2.00 (1)	2.00 (1)	2.00 (3)	2.50 (4)
Venting	5.00 (6)	6.00 (5)	4.00 (4)	2.00 (4)
Positive reframing	6.00 (6)	5.50 (4)	5.00 (5)	5.50 (3)
Planning	7.00 (5)	6.00 (5)	7.00 (4)	4.50 (5)
Humour	4.00 (6)	4.00 (6)	2.50 (3)	3.00 (2)
Acceptance	6.00 (5)	6.00 (4)	6.00 (6)	6.00 (6)
Religion	4.00 (6)	4.00 (3)	4.00 (5)	4.00 (3)
Self-blame	4.00 (4)	3.00 (2)	2.50 (4)	2.50 (2)

Note: M: median; n: number of participants; BCOPE: German version of the brief COPE tool for coping styles

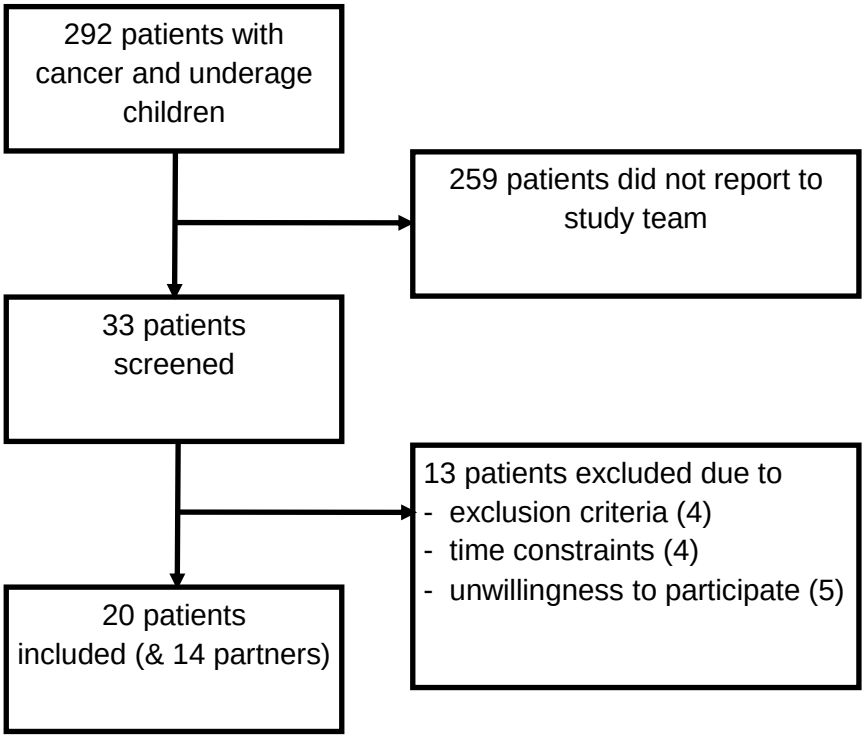

Figure 1: Inclusion